

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	A Study Protocol for a Randomized Clinical Trial of a Decision Aid and Values Clarification Method for Parents of a Fetus or Neonate Diagnosed with a Life-Threatening Congenital Heart Defect
AUTHORS	Delaney, Rebecca; Pinto, Nelangi; Ozanne, Elissa; Stark, Louisa; Pershing, Mandy; Thorpe, Alistair; Witteman, Holly; Thokala, Praveen; Lambert, Linda; Hansen, Lisa; Greene, Tom; Fagerlin, A

VERSION 1 – REVIEW

REVIEWER	Gendler, Yulia Ariel University
REVIEW RETURNED	27-Jul-2021

GENERAL COMMENTS	<p>The authors are congratulated for dealing with this sensitive and complex issue and on writing a clear and detailed research protocol. The paper is well written. It may be further improved by trying to deal with some of the following minor issues, if possible.</p> <p>General comments:</p> <p>The protocol does not clearly specify at what stage the clinicians are involved. For example, what is the natural course of the counseling process following prenatal diagnosis congenital heart disease in the control group? Is there a conversation with the clinician immediately after the diagnosis? At what stage does this conversation happens in research groups? How are you dealing with possible unrecognized bias on the part of the clinician?</p> <p>Specific comments:</p> <p>1. Table 1 – Decision Aid Content: 'How We Talk About Congenital Heart Defects': When introducing parents to topics and terms that are often used when discussing congenital heart defects, it is worth adding discussion regarding possible neurological complications.</p> <p>2. Table 2 – Use of Information Sources: Consider adding to your list 'consultation with a spiritual advisor' and 'seek for a second opinion'.</p> <p>3. Table 2 – Demographics and Religiosity: Religiosity is a complex construct and DUREL index serves as a good fit for measuring it. However, you are using a brief version with only 2 items out of 5. Level of religiosity found to be a major factor that influences parents' decision making (a few articles on this specific topic have been recently published). Please consider using</p>
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	<p>all 5 items or adding a direct question such as 'how do you define yourself: secular / traditional / religious / orthodox?'. 4. Data analysis: Decision regarding the fate of the pregnancy / the newborn is a shared decision making involving both spouses. Please consider adding dyadic data analysis to your data analysis plan.</p>
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REVIEWER	Gowda, Sharada Baylor College of Medicine
REVIEW RETURNED	24-Aug-2021

GENERAL COMMENTS	A very important topic. I applaud the authors/researchers in selecting a topic with such complexity both medically and psychosocially.
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VERSION 1 – AUTHOR RESPONSE

Reviewer: 1
Dr. Yulia Gendler, Ariel University

Comments to the Author:

The authors are congratulated for dealing with this sensitive and complex issue and on writing a clear and detailed research protocol. The paper is well written. It may be further improved by trying to deal with some of the following minor issues, if possible.

Response: Thank you very much for your review and support of our study. We greatly appreciate your feedback on how to strengthen our protocol.

General comments:

The protocol does not clearly specify at what stage the clinicians are involved. For example, what is the natural course of the counseling process following prenatal diagnosis congenital heart disease in the control group? Is there a conversation with the clinician immediately after the diagnosis? At what stage does this conversation happens in research groups? How are you dealing with possible unrecognized bias on the part of the clinician?

Response:

We have added the following statements to clarify the timing of counseling:

Patients consult with a clinician immediately after the diagnosis. Then, they are approached by research staff for study participation. (page 7, lines 8-9).

For the intervention groups, the decision aid is initiated by the parent, independent of the provider or coordinator. Both the control and intervention groups consult with clinicians as they decide which treatment to pursue. (page 7, lines 11-13).

Our study analyses will account for variation in outcomes across providers (see page 14-15) when making statistical inferences. The focus of our intervention is on the patient and was designed, in part, to help prevent against issues related to physician bias. Other research studies could be conducted to identify and intervene upon potential clinician bias.

Specific comments:

1. Table 1 – Decision Aid Content:

'How We Talk About Congenital Heart Defects': When introducing parents to topics and terms that are often used when discussing congenital heart defects, it is worth adding discussion regarding possible

neurological complications.

Response: We do highlight developmental delays in mental and cognitive abilities as part of something to consider related to quality of life. We have added this example within Table 1.

2. Table 2 – Use of Information Sources:

Consider adding to your list 'consultation with a spiritual advisor' and 'seek for a second opinion'.

Response: Thank you for this suggestion. We have amended the measure to include 'spiritual or religious advisor.' We did not include seek a second opinion as this would be captured under the 'providers' item. Our intention with this question is to better understand the type of people or resources people tend to use for their information.

3. Table 2 – Demographics and Religiosity:

Religiosity is a complex construct and DUREL index serves as a good fit for measuring it. However, you are using a brief version with only 2 items out of 5. Level of religiosity found to be a major factor that influences parents' decision making (a few articles on this specific topic have been recently published). Please consider using all 5 items or adding a direct question such as 'how do you define yourself: secular / traditional / religious / orthodox?'

Response: We agree that religiosity is an important factor to account for in regards to parental decision making in this context. We have a question included in our survey that we believe is similar to the direct question you suggest where we ask participants, "what is your present religion, if any?" with the following response options: Protestant, Roman Catholic, Latter Day Saint, Orthodox, Jewish, Muslim, Buddhist, Hindu, Atheist (you believe there is no God), Agnostic (you are not sure if there is a God),

Spiritual, but not committed to a particular faith, Something else, Nothing in particular. Therefore, we will not include additional questions in the interest of minimizing survey burden.

4. Data analysis:

Decision regarding the fate of the pregnancy / the newborn is a shared decision making involving both spouses. Please consider adding dyadic data analysis to your data analysis plan.

Response: Thank you for this suggestion. In our mixed effects models, we plan to examine family effects (parent dyad) to assess parent variation (pages 14-15). We also added that if we have sufficient power we will examine parent dyads in a sub-analysis (page 13, line 22): "If there is sufficient power to detect differences, exploratory sub-group analyses may be conducted to detect differences by factors such as pre versus post-natal diagnoses, CHD diagnosis, provider specialty, and parent dyads."

Reviewer: 2

Dr. Sharada Gowda, Baylor College of Medicine

Comments to the Author:

A very important topic. I applaud the authors/researchers in selecting a topic with such complexity both medically and psychosocially.

Response: Thank you very much for your review. We greatly appreciate your support of our study.

Reviewer comments from attached file:

-Page 6, lines, 24-29: May perceive: this statement is ambiguous. Do they mean though there is no equipoise the medical team may implicitly assume that there is equipoise?

Response: In consideration of the lack of clarity regarding this sentence, we have removed the statement from the paper.

-Page 9, lines 27-29: Curious to know if this resonates with decision making differences between parents later, as in if one wants the tool and other declines, would they arrive at different decision or does arrival point is prolonged?

Response: We agree that this would be an interesting question to explore. We may not be able to directly address this, however, because in our study it is possible that both parents view the decision aid (only 1 tablet is given per family) and only 1 parent completes the survey to be included in the

study. In fact, currently all parents have indicated they have reviewed the decision aid with their partner, even if only one parent completes the surveys. Please also see our response to Reviewer 1, #4 about conducting dyadic analyses, which we believe may also be relevant to this comment.